

Original Article

Long-term enteral feeding and gastroesophageal reflux disease in neurologically impaired children

P. DE ANGELIS¹, A. PANE¹, T. CALDARO¹, F. TORRONI¹, G. FEDERICI DI ABRIOLO¹, G. CIOFETTA², I. ALOI¹, A. CROCOLI¹, F. FOSCHIA¹, L. DALL'OGGIO¹

¹Digestive Surgery and Endoscopy Unit, Pediatric Hospital Bambino Gesù, IRCCS, Rome

²Nuclear Medicine Unit, Pediatric Hospital Bambino Gesù, IRCCS, Rome - Italy

ABSTRACT: Background. In neurologically impaired children, feeding problems and gastroesophageal reflux disease require great care throughout the children's lifetimes. It is difficult to decide on the right time for diagnostic-therapeutic procedures. A vicious circle between gastroesophageal reflux disease, swallowing incoordination, and problems with aspiration leads to severe malnutrition, with poor quality of life.

Objective. To retrospectively review a selected population and describe our management of these patients.

Patients/methods. Neurologically impaired, malnourished patients (n = 261) underwent triceps skinfold thickness measurement, endoscopy/biopsy, and/or 24-hour pH-metry, videofluoroscopy-salivagram, upper gastrointestinal series, and gastric scintigraphy. The therapeutic options were chosen by a multidisciplinary team: percutaneous endoscopic gastrostomy, swallowing rehabilitation, Nissen fundoplication, pyloromyotomy (in delayed gastric emptying), total esophagogastric disconnection, EndoCinch, and/or gastrojejunal tube.

Results. Surgical/endoscopic interventions chosen were 217 percutaneous endoscopic gastrostomies, 25 Nissen fundoplications/gastrostomies, 16 pyloromyotomies, 15 primary total esophagogastric disconnections, and 4 EndoCinch procedures. No operative mortality occurred; postoperative courses showed few complications. In the follow-up treatment, symptoms improved as did quality of life.

Conclusion. Neurologically impaired children with gastroesophageal reflux disease, eating inability and malnutrition should be treated with rehabilitation planning, precocious gastrostomy, and early therapy for gastroesophageal reflux disease. (*Nutritional Therapy & Metabolism* 2007; 25: 147-52)

KEY WORDS: Enteral feeding, Gastroesophageal reflux disease (GERD), Neurologically impaired children (NIC)

INTRODUCTION

In neurologically impaired children (NIC), feeding problems and gastroesophageal reflux disease (GERD) are widespread and require great care throughout their lifetimes.

It is often difficult to choose and decide on the right time for each diagnostic and therapeutic procedure, because of doubts regarding the reality of the unfavorable patient prognosis and parents' expectations. Generally,

an unpleasant vicious circle is established (Fig. 1) between GERD, swallowing incoordination, and difficulties with aspiration (*dyscatoposia*) in NIC. With the addition of constipation, the final result is severe malnutrition with a high susceptibility to infection and a poor quality of life, and consequent difficulty in management of every daily activity by parents and caregivers (1).

An accurate diagnosis of GERD in NIC is not always easy, because there may be no clear symptoms. Convulsions and hyper-tone resistant to neurologic

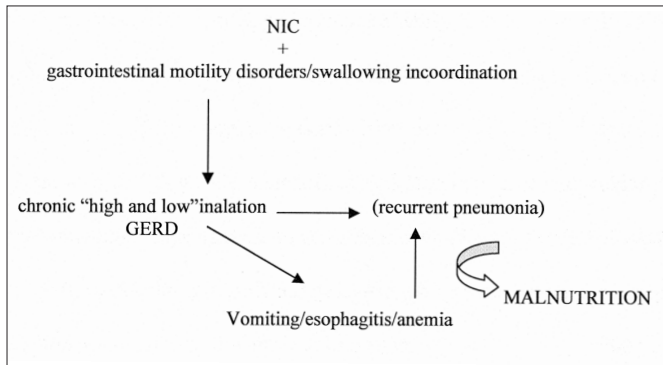


Fig. 1 - The pathologic circle in neurologic impairment. GERD = gastroesophageal reflux disease; NIC = neurologically impaired children.

drugs, chronic cough, and recurrent *ab ingestis* pneumonia are frequently signs of severe GERD, as are, obviously, severe anemia, melena, and anorexia. More than half of the reflux events in NIC are nonacidic and would therefore go undetected by conventional pH-metry (2).

Gastric feeding through a nasogastric tube is often required in NIC when oral intake is not satisfactory, but in long-term follow-up a gastrostomy is essential to ensure good nutrition without difficulty. Percutaneous endoscopic gastrostomy (PEG) is a safe and effective procedure, including in patients severely disabled with substantial anatomic deformities and restricted pulmonary function; it has become the procedure of choice for the establishment of enteral access in patients requiring long-term enteral alimentation (3).

Oral rehabilitation should be added to enteral feeding and included in a multidisciplinary approach involving nurses, therapists, and dieticians, in addition to gastroenterologists, neurologists, and surgeons. Medical therapy and innovative endoscopic and surgical techniques are necessary to manage GERD (1). Symptomatic GERD can be treated with proton pump inhibitors (PPIs) until it is impossible to control the symptoms and signs, and antireflux surgery becomes absolutely necessary (4). Nissen fundoplication is considered the best surgical option in patients affected by gastroesophageal reflux disease (5). Nevertheless, NICs have a high risk of gastroesophageal reflux disease recurrence after fundoplication because of several negative factors. Total esophagogastric dissociation (TEGD) eliminates all risk of recurrent reflux and can be used as a primary treatment of choice for severely NIC completely dependant on tube feeding, with significant oropharyngeal incoordination (6, 7). In very compromised NIC patients, with a high surgical and anesthesiologic risk, recurrent vomiting, and severe malnutrition, a gastrojejunal tube can

easily be placed even with some limitations, if a gastrostomy has been previously performed. Although gastrojejunal feeding tubes can be inserted safely and improve nutritional status, the most common complication is jejunal tube migration with gastric dislocation (8). Duodenostomy or jejunostomy are invasive and more definitive procedures, with high incidence rates of complications.

The aim of our study was to retrospectively review a selected NIC population, describing our clinical approach to the patients' diagnostic and therapeutic management.

PATIENTS AND METHODS

In a retrospective analysis, we reviewed the clinical cards of 261 patients (151 males). All patients were severely NIC according to the criteria at that time of the American Psychiatric Association (9). All patients presented severe malnutrition (triceps skinfold thickness < fifth percentile) and GERD from mild to severe degree; they underwent nutritional intervention from 1992 to 2006. Diagnoses were postasphyxial encephalopathy in 204 patients (189 postpartum, 2 post-heart surgery, 4 postmeningitis, 3 postdrowning, 6 post-traumatic asphyxia), genetic and chromosomal syndrome in 52 patients, and metabolic disease in 5 patients. Mean age at surgical/endoscopic intervention (PEG or antireflux surgery) was 8 years and 5 months (range 1 months to 20 years); 256 patients (98%) were < 10 years old, 5 of them < 6 months. Weight of patients ranged from 3.3 to 48 kg; for patients 10-20 years old, mean weight was 21.3 kg (range 10.8-48 kg); for patients 6 months to 10 years old, mean weight was 10.5 kg (range 5.5-14.8 kg); and for patients < 6 months, mean weight was 4.3 kg (range 3.3-5.2 kg). All patients presented severe malnutrition, defined as triceps skinfold thickness below the fifth percentile for age and sex (10), and required nasogastric tube feeding for more than 3 months; they did not have evident gastrointestinal alterations.

Diagnostic steps

- To assess malnutrition: triceps skinfold thickness;
- To document GERD: positive endoscopy with biopsy (peptic esophagitis) or positive 24-hour pH-metry, showing atypical symptomatology and mild or no esophagitis;
- In suspected cases of high aspiration: contrast swallow with videofluoroscopy (11) or/and salivagram (12);
- To rule out peptic esophageal stricture and to detect gastrointestinal malformation before PEG placement:

upper gastrointestinal series;

- To evaluate quantitatively gastric emptying and to decide on the best surgical procedure in GERD: scintigraphy (13).

Therapeutic steps

- Patients with inadequate intake, with a positive response to enteral feeding through nasogastric tube, without severe GERD or with GERD medically controllable, and patients who were in too poor a general condition to be operated on underwent only PEG with swallowing rehabilitation and PPI;
- Patients with GERD resistant to drugs with partial oral feeding underwent Nissen fundoplication (open or laparoscopic according to the patient's situation) with gastrostomy (if no previous PEG) and pyloromyotomy (in delayed gastric emptying as evaluated by scintigraphy);
- Patients with recurrence of GERD after Nissen fundoplication or patients dependent on enteral feeding without oral intake or with severe dyscatoposia underwent TEGD (*rescue or primary*) with gastrostomy (if no previous PEG) and pyloromyotomy (in delayed gastric emptying as evaluated by scintigraphy).
- Patients with mild/moderate GERD dependent on drugs, with acceptable oral feeding, no high hiatal hernia, prevalently respiratory symptoms, and regular gastric emptying underwent EndoCinch gastroplication.
- Patients with severe recurrent vomiting and retching, with a very high surgical and anesthesiologic risk and previous gastrostomy, underwent insertion of gastrojejunal tube with or without baclofen; in cases of instability of gastrojejunal tube and very poor quality of life, a jejunostomy was performed.

RESULTS

The results are summarized in Figure 2. Therapeutic interventions were as follows:

- 217 patients underwent PEG; afterwards, 25 of them, Nissen fundoplication post-PEG (15, pyloromyotomy; 6, TEGD post-antireflux surgery as a rescue procedure after GERD recurrence for Nissen fundoplication disruption); 5 of them, TEGD post-PEG; 3 of them, EndoCinch trans-PEG (with 1 Nissen fundoplication + pyloromyotomy post-EndoCinch, 1 TEGD post-EndoCinch); 30 patients underwent 42 gastrojejunal tube (2 jejunostomy); and 154 patients are still on PPIs and neurologic drugs;
- 25 underwent Nissen fundoplication + gastrostomy +

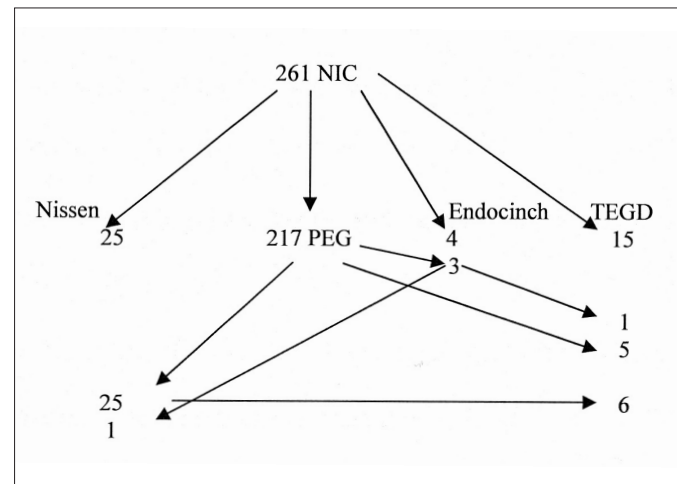


Fig. 2 - Therapeutic options chosen for patient population ($n = 261$). PEG = percutaneous endoscopic gastrostomy; TEGD = total esophagogastric dissociation.

- pyloromyotomy (16 patients);
- 15 underwent TEGD primary and gastrostomy + pyloromyotomy;
- 4 underwent EndoCinch gastroplication.

In all patients except one, enteral feeding was started through gastrostomy (PEG or gastrostomy placed during Nissen fundoplication or TEGD) either for nutritional reasons or to improve quality of life and alleviate respiratory symptoms: in the result, 125 patients had unsatisfactory oral feedings with long duration of meals and constant cough during uncoordinated swallowing. The rest of them, 136 patients, had a permanent nasogastric tube placed a mean of 4 months before (range 1-7 months) with improved efficacy of daily and nightly enteral feeding (no weight loss reported by parents) but persistence of chronic bronchitis, perhaps related to microinhalations. In 67 patients with PEG, parents decided on exclusive enteral feeding by gastrostomy due to recurrent "high" levels of microinhalation. Oral rehabilitation is still continuing with success in 65 patients.

There was no operative mortality, and postoperative courses presented only a few complications related to surgical procedures:

- 5 gastrocolic fistulas post-PEG, surgically treated;
- 2 cases of hemorrhage immediately post-EndoCinch with spontaneous resolution;
- 2 cases of bowel obstruction (1 adhesion, 1 caecum volvulus) and 2 esophagojejunal anastomotic stenoses (cicatrical peptic stricture resolved by dilatation), post-rescue TEGD;
- 1 tight fundoplication, which required hydrostatic dilation.

Follow-up treatment, which ranged from 1 year to 12 years, was not a specific topic of our examination. Eight patients died due to underlying disease.

The general health of patients after nutritional intervention and antireflux surgery improved, with regard to the number of episodes of food aspiration, cough, and respiratory infections (reduction in 118 patients from a mean of 21 to 8, as evaluated after the first year post-operative therapy; 77 of these patients underwent a Nissen fundoplication or TEGD); gastrointestinal symptoms, such as vomiting and regurgitation observed (disappearance in 98 patients surgically treated); chronic anemia (resolved in 7 patients); and mean relative weight (improved in 215 patients with increase of body weight from 1.5 to 6 kg after the first year post-operative therapy). Also, the satisfaction of parents, as reported during the semester check, testified to a better quality of life, specially after PEG or gastrostomy.

The persistence of retching in 150 patients (48 out of whom had undergone surgical correction for GERD) represented the most important problem reported by parents during the follow-up.

DISCUSSION

As the literature shows, there is no common agreement on how to manage NIC either in terms of diagnosis or therapy; however, it is necessary to find a homogeneous route.

Regarding diagnosis of GERD, the recent, still not widespread use of combined 24 hours pH-metry and multiple intraluminal impedance allows the detection of both acid (pH < 4) and nonacid (pH > 4) GER episodes, with a better diagnosis overall in NIC (2).

Upper digestive tract scintigraphy, with a salivagram is a rarely utilized exam important to detect high aspiration aside from GERD. Chronic salivary aspiration may be responsible for pneumonia in NIC; radionuclide salivagram is effective in documenting salivary aspiration as the source of recurrent pneumonia (12, 14). According to Baikie et al, the maximum agreement is between aspiration as diagnosed with salivagram and by barium fluoroscopy (15).

With regard to therapeutic interventions against malnutrition, there is no doubt that the indication for gastrostomy is overall inadequate oral nutrition; certainly, gastrostomy can avoid the higher aspiration levels caused by swallowing incoordination, but it cannot prevent GERD. PEG can be performed in a brief time on general or local anesthesia, with rare major complications; when it is possible, the gastrostomy should be placed in the lesser gastric curvature, because of demon-

strated antirefluxing properties (16). Besides, PEG is useful to improve malnutrition and the general condition of patients, providing effective access for long-term enteral nutrition (3).

According Lewis et al, successful nutritional rehabilitation can improve GERD and can be achieved in malnourished NIC, using PEG feeding and antireflux medication, even if some NIC may need antireflux surgery (10).

A child neurologist should be involved in the multidisciplinary oromotor rehabilitation program to improve swallowing difficulties and their consequences (11). When there is a clear failure of medical therapy, very poor quality life, frequent gastrojejunal tube dislocations, and constant aspiration pneumonia, then early antireflux surgery is suggested.

Nissen fundoplication remains the standard technique for correction of GERD, including in NIC, resolving major respiratory symptoms in particular (5, 17, 18). Nissen fundoplication is considered the best surgical option in patients affected by GERD, but in NIC, it is burdened with retching, an early component of the emetic reflex. The γ -aminobutyric acid type B receptor agonist baclofen reduces the incidence of transient lower esophageal sphincter relaxation; and repeated administration of baclofen reduces the frequency of emesis and the total number of acid refluxes in NIC (19).

We often perform a gastrostomy before antireflux surgery, to improve nutritional status and to have fewer surgical complications. Other investigators have also changed their approach, regardless of pH study results, managing GERD pharmacotherapeutically and reserving surgery for cases where this proves inadequate (4).

NIC have a high risk of GERD recurrence after fundoplication; the options available after failed Nissen fundoplication are jejunal feeding through a jejunostomy or a nasojejunal or gastrojejunal tube (8). To eliminate one of the factors involved in recurrence of GERD, treatment of documented delayed gastric emptying has been proposed through pyloromyotomy or pyloroplasty. A retrospective study by Burstoff Silva et al confirmed the importance of pyloromyotomy to avoid the recurrence of GERD (20). Comparing scintigraphy pyloromyotomy with pyloroplasty in a long-term follow-up of patients who had undergone antireflux surgery, Okuyama et al described fewer complications in the postoperative course of the pyloromyotomy group (21). Besides, pyloromyotomy is easier to perform and associated with less morbidity.

Nevertheless, when in NIC the conventional antireflux surgery has failed, it is necessary to find other options. Redo fundoplication has a higher recurrence rate (25%-50%) than the initial operation and carries a

greater risk of complications (22, 23). A very interesting option is TEGD which eliminates all risk of recurrent reflux and can be used as a primary treatment of choice for severely NIC completely dependent on tube feeding with significant oropharyngeal incoordination (6, 7). A previous study by our group demonstrated that TEGD offers better nutritional rehabilitation, reduction in respiratory infections, and improved quality of life, compared with fundoplication and gastrostomy (24, 25).

It is obviously very important to effect an accurate selection of patients undergoing TEGD, evaluating the real neurologic damage, the degree of dyscatoposia, and the family acceptance of a radical surgical option.

Lastly, we would mention several new, less-invasive endoscopic techniques for GERD, which are safe and effective also in children and adolescents (26). We have used endoscopic gastroplication with an EndoCinch device, as well as through gastrostomy, with partially positive results. Further improvement of the procedure, applicable to NIC, and more trials are necessary to obtain better success (27).

CONCLUSION

Children with profound neurologic impairment caused by heterogeneous factors which lead to a persistent vegetative state with consequent GERD, inability to eat orally, and severe malnutrition, can be treated with a careful planning of rehabilitation, to improve the quality of life of the patients and their caregivers. Precocious gastrostomy results in a better nutritional status and the possibility of confronting all of the difficult complications of underlying disease. A good knowledge and ear-

ly diagnosis and treatment of GERD may help these NIC patients to avoid a number of unpleasant complications which lead to a poor life prognosis. Early involvement by a multidisciplinary team of physicians, nurses, dieticians, occupational and speech therapists, psychologists, and social workers is essential to prevent the adverse outcomes associated with feeding difficulties and poor nutritional status. Careful evaluation and monitoring of severely disabled children for nutritional problems are warranted because of the increased risk of nutrition-related morbidity and mortality (28).

Further, GERD and malnutrition are certainly the major determinants of perioperative morbidity and mortality in NIC. The incidence of these risk factors can be reduced through careful preoperative evaluation and nutritional rehabilitation (29).

ACKNOWLEDGEMENTS

We thank Mrs Maria Bettini and Mrs Stella Fiorenza from our Digestive Surgical and Endoscopic Unit for their daily kind help and support.

Conflict of interest statement

None declared.

Address for correspondence:
Paola De Angelis, MD
Via Cassia, 1280
00189 Rome, Italy
e-mail paolade.angelis@tiscali.it,
paola.deangelis@opbg.net

REFERENCES

1. Chong SK. Gastrointestinal problems in the handicapped child. *Curr Opin Pediatr* 2001; 13: 441-6.
2. Del Buono R, Wenzl TG, Rawat D, Thomson M. Acid and nonacid gastro-oesophageal reflux in neurologically impaired children: investigation with the multiple intraluminal impedance procedure. *J Pediatr Gastroenterol Nutr* 2006; 43: 331-5.
3. Ponsky JL, Gauderer MW, Stellato TA, Aszodi A. Percutaneous approaches to enteral alimentation. *Am J Surg* 1985; 149: 102-5.
4. Wilson GJ, van der Zee DC, Bax NM. Endoscopic gastrostomy placement in the child with gastro-esophageal reflux: is concomitant antireflux surgery indicated? *J Pediatr Surg* 2006; 41: 1441-5.
5. Georgeson K. Results of laparoscopic antireflux procedures in neurologically normal infants and children. *Semin Laparosc Surg* 2002; 9: 172-6.
6. Morabito A, Lall A, Lo Piccolo R, et al. Total esophago-gastric dissociation: 10 years' review. *J Pediatr Surg* 2006; 41: 919-22.

7. Lall A, Morabito A, Dall'Oglio L, et al. Total oesophago-gastric dissociation: experience in 2 centres. *J Pediatr Surg* 2006; 41: 342-6
8. Godbole P, Margabanthu G, Crabbe DC, et al. Limitations and uses of gastrojejunal feeding tubes. *Arch Dis Child* 2002; 86: 134-7.
9. Spitzer RL, Williams JBW, eds. Diagnostic and statistical manual of mental disorders; 3rd ed. Washington, DC: American Psychiatric Association, 1987.
10. Lewis D, Khoshoo V, Pencharz PB, Golladav ES. Impact of nutritional rehabilitation on gastroesophageal reflux in neurologically impaired children. *J Pediatr Surg* 1994; 29: 167-9.
11. Fung CW, Khong PL, To R, Goh W, Wong V. Videofluoroscopy study of swallowing in children with neurodevelopmental disorders. *Pediatr Int* 2004; 46: 26-30.
12. Cook SP, Lawless S, Mandell GA, Reilly JS. The use of the salivagram in the evaluation of severe and chronic aspiration. *Int J Pediatr Otorhinolaryngol* 1997; 41: 353-61.
13. Malmud LS, Fisher RS. Scintigraphic evaluation of disorders of the esophagus, stomach, and duodenum. *Med Clin North Am* 1981; 65: 1291-310.
14. Heyman S. The radionuclide salivagram for detecting the pulmonary aspiration of saliva in an infant. *Pediatr Radiol* 1989; 19: 208-9.
15. Baikie G., South MJ, Reddihough DS, et al. Agreement of aspiration tests using barium videofluoroscopy, salivagram, and milk scan in children with cerebral palsy. *Dev Med Child Neurol* 2005; 47: 86-93.
16. Stringel G. Gastrostomy with antireflux property. *J Pediatr Surg* 1990; 25: 1019-21.
17. Al-Mohaidaly M, Steyaert H, Valla JS. Long term results of laparoscopic antireflux surgery in children. *Saudi Med J* 2003; 24 (5 Suppl): S29.
18. Leon FF, Gasparella M, Gamba PG. Antireflux surgery in neurologically impaired children. *Pediatr Med Chir* 2005; 27: 75-80.
19. Kawai M, Kawahara H, Hirayama S, Yoshimura N, Ida S. Effect of baclofen on emesis and 24-hour esophageal pH in neurologically impaired children with gastroesophageal reflux disease. *J Pediatr Gastroenterol Nutr* 2004; 38: 317-23.
20. Burstoff Silva J, Fonkalsrud EW, Perez CA, et al. Gastric emptying procedures decrease the risk of postoperative recurrent reflux in children with delayed gastric emptying. *J Pediatr Surg* 1999; 34: 79-82.
21. Okuyama H, Urao M, Coran G, et al. A comparison of the efficacy of pyloromyotomy and pyloroplasty in patients with gastroesophageal reflux and delayed gastric emptying. *J Pediatr Surg* 1997; 32: 316-20.
22. Buratti S, Kamenwa R, Dohil R, Collins D, Lavine JE. Esophagogastric disconnection following failed fundoplication for the treatment of gastroesophageal reflux disease (GERD) in children with severe neurological impairment. *Pediatr Surg Int* 2004; 20: 786-90.
23. Wheatley MJ, Coran AG, Wesley JR, et al. Redofundoplication in infants and children with recurrent gastroesophageal reflux. *J Pediatr Surg* 1991; 26: 758-61.
24. Gatti C, Federici di Abriola G, Villa M, et al. Esophagogastric dissociation versus fundoplication: which is the best for severely neurologically impaired children? *J Pediatr Surg* 2001; 36: 677-80.
25. Danielson PD, Emmens RW. Esophagogastric disconnection for gastroesophageal reflux in children with severe neurological impairment. *J Pediatr Surg* 1999; 34: 84-6.
26. Thomson M, Fritscher-Ravens A, Hall S, Afzal N, Ashwood P, Swain CP. Endoluminal gastroplication in children with significant gastro-oesophageal reflux disease. *Gut* 2004; 53: 1745-50.
27. Schwartz MP, Wellink H, Gooszen HG, Conchillo JM, Samsom M, Smout AJ. Endoscopic gastroplication for the treatment of gastro-oesophageal reflux disease: a randomised, sham-controlled trial. *Gut* 2007; 56: 20-8.
28. Marchand V, Motil KJ; NASPGHAN Committee on Nutrition. Nutrition support for neurologically impaired children: a clinical report of the North American Society for Pediatric Gastroenterology, Hepatology, and Nutrition. *J Pediatr Gastroenterol Nutr* 2006; 43: 123-35.
29. Glassman MS, Dozer AJ, Newman LJ. Gastroesophageal reflux in neurologically impaired children: perioperative evaluation and management. *South Med J* 1992; 85: 289-92.

Received: November 22, 2006

Accepted: August 28, 2007